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RECURRENT KERATOCYSTIC ODONTOGENIC TUMOUR - A 17-YEAR FOLLOW-UP CASE REPORT

Medicine

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ABSTRACT

Keratocystic odontogenic tumour is eccentric among the cyst of the maxillofacial region. In spite of generous research, the universally accepted treatment is still debated. We report a case of Keratocystic odontogenic tumour in a 35-year old male who had a recurrence within three and a half years of conservative treatment. Hence during the second time, the aggressive treatment protocol was carried out. There was no recurrence noted after 17 years follow-up.

KEYWORDS

Odontogenic keratocyst, Keratocystic odontogenic tumour, multilocular radiolucency, aggressive treatment, enucleation, Carnoy's solution

INTRODUCTION:

Odontogenic keratocyst (OKC) was first described by Philipsen in 1956 [1]. which now is designated by the World Health Organization (WHO) as Keratocystic odontogenic tumour (KCOT) [2] due to its aggressive nature of growth, post-treatment recurrence and an association with nevoid basal cell carcinoma syndrome [3]. The highest incidence is between 10 and 30 years of age, favouring the male population in the posterior part of the body and ramus of mandible [4]. Various conservative and aggressive therapies are available in the literature, but yet to decide the universally accepted treatment. However, cyst eradication, reduction of the recurrence and surgical morbidity are the ultimate goals of all the techniques. We, therefore, present a case of recurrent KCOT in which aggressive treatment was executed with no recurrence after a follow-up of 17 years. Thus, adds strong evidence to the literature.

Case History:

A 35-year old male patient reported to a dental college in the year 2004 with the complaint of swelling in the lower front teeth region for the past two months. The swelling was initially small sized which gradually increased to its present size. It was associated with pain for the past 10 days. There were no other associated symptoms. Past dental history revealed that the patient had a similar kind of symptoms (Figure 1) three and a half years back and was diagnosed as KCOT for which enucleation was done in our dental college.

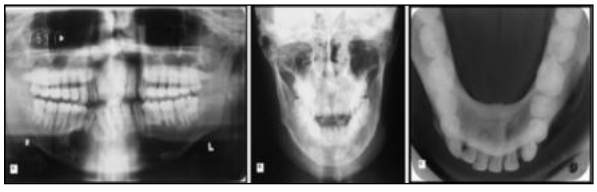

Figure 1: Radiographs Shows KCOT At The Time Of Initial Presentation.

Past medical history, general examination and extraoral examination revealed no abnormalities (Figure 2 A). On intraoral soft tissue examination, diffuse swelling was seen in vestibular mucosa in 34, 35 and in 42, 43 region of approximate size 1x1.5 cm and 0.5x1.0 cm respectively (Figure 2 B and C) which was hard in consistency and tender.

Buccal cortical plate expansion was present in 34, 35 region. Hard tissue examination revealed missing 43, distally tilted 42. Considering the history and clinical findings, a provisional diagnosis of recurrent KCOT was given. All the lower anteriors were vital. White viscous fluid was aspirated (Figure 3) and the cytopathological features suggested KCOT.

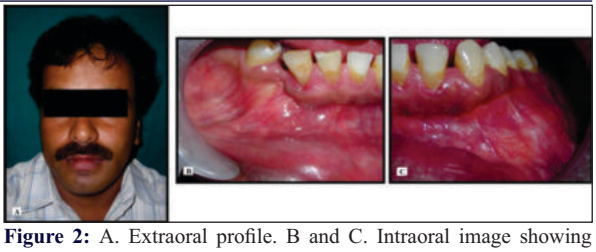

Figure 2: A. Extraoral profile. B and C. Intraoral image showing diffuse swelling in vestibular mucosa in 34, 35 and 42, 43 regions.

Figure 3: Aspiration Showing White Viscous Fluid.

Complete haemogram and chest X-ray was normal, OPG (orthopantomogram) and PA (posteroanterior) mandible showed a well-defined multilocular radiolucency of size 6x2.5 cm approximately present in the periapical region of 36 to 44. Buccal cortical plate expansion was seen in 34, 35 and 45, 46 regions on the mandibular occlusal radiograph (Figure 4 A, B and C).

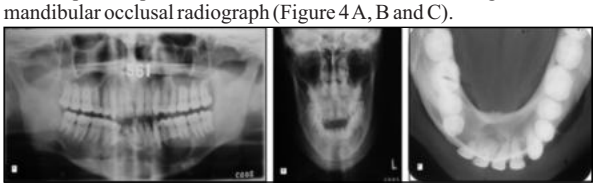

Figure 4: A and B. Radiographs shows KCOT at the time of recurrence. A well-defined multilocular radiolucency in the periapical region of 36 to 44 with C. Buccal cortical plate expansion in 34, 35 and 45, 46 region.

Surgery was planned under General Anaesthesia, the arch bar was placed in the upper and lower arches, a vestibular incision was given and full thickness mucoperiosteal flap was elevated, the area of bony expansion was identified, bone guttering done followed by enucleation with curettage supplemented with Carnoy's solution application.

Histopathology report showed parakeratinised corrugated stratified squamous epithelium of 6-8 cell layer thickness having no rete ridges and palisaded and polarised cuboidal cells in the basal cell layer (Figure 5).

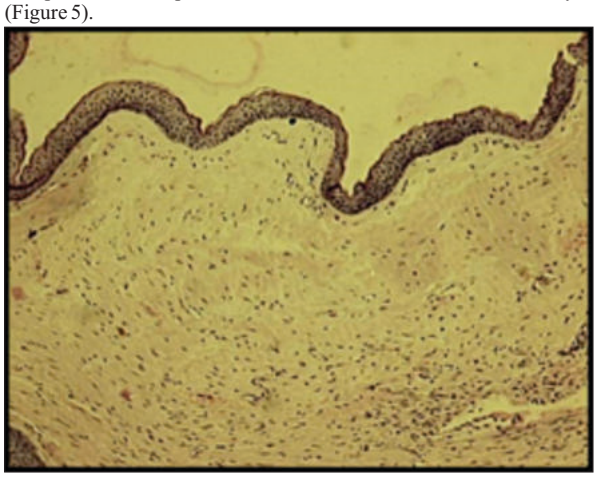

Figure 5: Histopathological image showing para keratinised corrugated stratified squamous epithelium of 6-8 cell layer thickness without any rete ridges and palisaded and polarised cuboidal cells in the basal cell layer.

Post-operative radiographs were taken immediately, three months and seven months later (Figure 6).

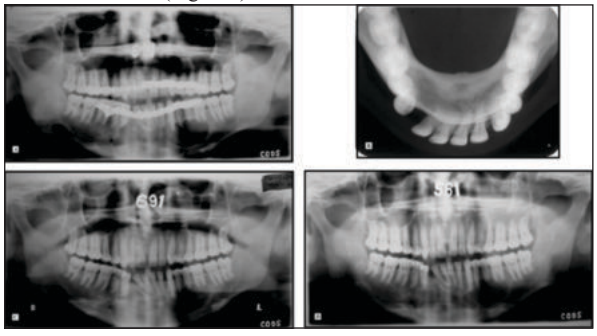

Figure 6: Post-operative radiographs A. Immediate post-op. B and C. Three months post-op. D. Seven months post-op.

Clinically, there was no swelling or obliteration in the vestibular mucosa after a month post-operatively (Figure 7).

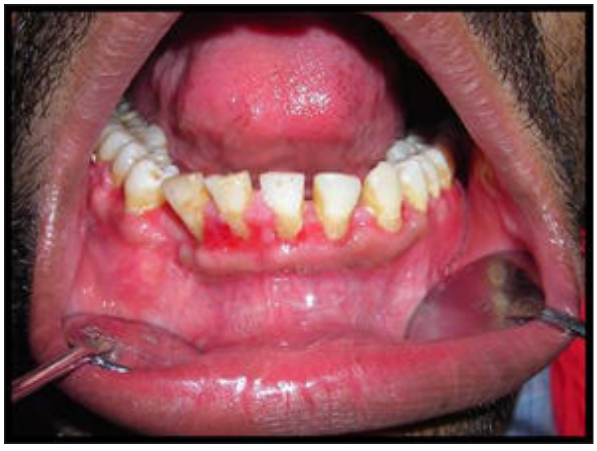

Figure 7: Intraoral image showing no swelling or obliteration in the vestibular mucosa after a month post-operatively.

The patient was followed-up every year through a telephonic interview. The patient was symptom-free until the time of write-up.

DISCUSSION:

KCOT is a well-known entity for its aggressive growth and recurrence [5]. The patient may present with pain, swelling, discharge, paraesthesia or maybe asymptomatic [6]. Radiographically it presents as well-defined uni or multilocular radiolucency and in a majority of the situation is confused with other odontogenic or non-odontogenic tumours. A unique feature is the expansion in the medullary space in the anteroposterior direction rather than causing obvious expansion in bucco-lingual direction [7] and exhibiting heterogeneous fluid contents in T-2 weighted MRI images [8].

Various conservative therapies, like enucleation with or without curettage, and marsupialization to aggressive treatment modalities including peripheral ostectomy, cryotherapy with liquid nitrogen, Carnoy's solution application and resection [9] are available in the literature. As enucleation along with Carnoy's solution application is significantly better at eliminating recurrences [8], it was chosen as the best treatment option for our patient. As the parakeratinized variant has the aggressive growth potential and postoperative recurrence tendency due to increased mitosis, along with budding capacity in the basal layer in the lining epithelium [10]. the treatment outcome was assessed over 17 year period.

In recent years, plant-based alkaloid 'cyclopamine' a molecular mechanism-based drug has been formulated that may serve as a promising futuristic treatment option [11].

CONCLUSION:

Conservative management may be considered as the initial line of management. But due to the small number of reported cases and lack of follow-up data in the maxillofacial region, the recommended treatment of this disease should be made cautiously. Surgical intervention must be fraught with morbidity and recurrence.

REFERENCES:

1. Philipsen H. On keratocysts in the jaws. Tandlaegebladet 1956; 60:963–81.
2. L. Barnes, J. W. Eveson, P. Reichart, and D. Sindrasky, Eds. Keratocystic odontogenic tumor in WHO Classification of Tumors. Pathology and Genetics of Head and Neck Tumors 2005 IARC Press, Lyon, France, p. 306.
3. Jonathan Madras and Henry Lapointe. Keratocystic Odontogenic Tumour: Reclassification of the Odontogenic Keratocyst from Cyst to Tumour. JCDA 2008;74(2):165-165h.
4. H. Myoung, S. P. Hong, S. D. Hong et al. Odontogenic keratocyst: Review of 256 cases for recurrence and clinicopathologic parameters. Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontics 2001;91(3):328-333.
5. Chrcanovic B R and Gomez R S. Recurrence probability for keratocystic odontogenic tumots: An analysis of 6,427 cases. Journal of Cranio-Maxillofacial Surgery 2017;45:244-51.
6. Meara, JG.; Shah, S; Li, KK, Cunningham, MJ. The Odontogenic Keratocyst: A 20-Year Clinicopathologic Review. Laryngoscope 1998;108(2):280-3.
7. R. Boeddinghaus and A. Whyte. Current concepts in maxillofacial imaging. European Journal of Radiology 2008;66(3):396-418.
8. Güler N, Sençift K, Demirkol O. Conservative management of keratocystic odontogenic tumors of jaws. Scientific World Journal 2012. 2012 680397.
9. N. Blanas, B. Freund, M. Schwartz, and I. M. Furst. Systematic review of the treatment and prognosis of the odontogenic keratocyst. Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontics 2000;90(5):553-8.
10. A. Habibi, N. Saghravanian, M. Habibi, E. Mellati, and M. Habibi. Keratocystic odontogenic tumor: A 10-year retrospective study of 83 cases in an Iranian population. Journal of Oral Science 2007;49(3):229-35.
11. Zhang L, Sun ZJ, Zhao YF, Bian Z, Fan MW, Chen Z. Inhibition of SHH signaling pathway: molecular treatment strategy of odontogenic keratocyst. Med Hypotheses 2006;67(5):1242-4.